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Spontaneous rupture of common iliac artery associated with fibromuscular dysplasia presenting with colic pain suggestive of ureteral stone

AUTHOR(S):

Yoshioka, Iwao; Arichi, Naoko; Tokugawa, Shigeki; Kishikawa, Hidefumi; Nishimura, Kenji; Ichikawa, Yasuji

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SPONTANEOUS RUPTURE OF COMMON ILIAC ARTERY ASSOCIATED WITH FIBROMUSCULAR DYSPLASIA PRESENTING WITH COLIC PAIN SUGGESTIVE OF URETERAL STONE

Iwao Yoshioka, Naoko Arichi, Shigeki Tokugawa,
Hidefumi Kishikawa, Kenji Nishimura and Yasuji Ichikawa
The Departments of Urology, Hyogo Prefectural Nishinomiya Hospital

We report a case of spontaneous rupture of the common iliac artery associated with fibromuscular dysplasia (FMD). A 21-year-old previously healthy male presented with acute onset of colic pain, suspected to be caused by a ureteral stone. Abdominal computed tomography and angiography revealed a retroperitoneal hematoma caused by rupture of the common iliac artery. In spite of an emergency operation initiated quickly, the patient died. A pathological examination demonstrated FMD of the common iliac artery. Although very rare, it is important to bear in mind that the possibility of retroperitoneal hemorrhage exists in patient with sudden lumbago.

(Hinyokika Kiyo **53** : 721–724, 2007)

Key words : Fibromuscular dysplasia (FMD), Common iliac arterial rupture, Emergency angiography

INTRODUCTION

An isolated spontaneous rupture of the common iliac artery is rare, though cases have been reported in association with arteriosclerosis, Marfan syndrome, Ehlers-Danlos syndrome, and fibromuscular dysplasia (FMD)[1,2]. FMD, first described by Leadbetter and Burkland, is also recognized as a cause of renovascular hypertension[3,4]. The clinical manifestations of these diseases are found in others, as well, such as the carotid, brachial, coronary, mesenteric and iliac arteries. The affected artery tends to be primarily occlusive, while secondary events include aneurysm, dissection and finally rupture. We present a case of spontaneous rupture of common iliac artery associated with FMD and discuss the management.

CASE REPORT

A 21-year-old previously healthy male was presented with an acute onset of left abdominal pain and transient consciousness disorder. The family history showed vascular diseases in his father, who had rupture of a cerebral aneurysm, and older brother, who experienced rupture of a splenic arterial aneurysm. The patient was admitted to our hospital by ambulance with colic pain initially suspected of being caused by a ureteral stone. However, because the left side of the abdomen was hard showing swelling and the pain spread in a wide range, this condition was not typical colic pain caused by a ureteral stone and it was necessary to acquire more detailed information. Abdominal computed tomography revealed an extended retroperitoneal hematoma that centered on the periphery of the left kidney, extended to the periphery of the right kidney and spread into the pelvic cavity (Fig. 1). Based on these findings,

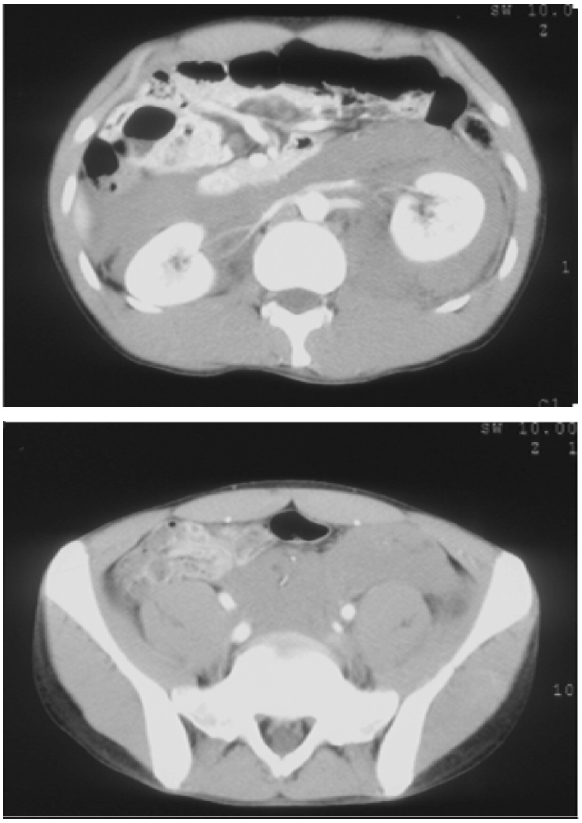

Fig. 1. Abdominal computed tomography images revealing an extended retroperitoneal hematoma.

emergency angiography was performed, because rupture of a left renal arterial aneurysm was suspected. Angiography of left renal artery demonstrated no obvious abnormalities, whereas subsequent aortagraphy revealed extravasation of the left common iliac artery (Fig. 2). Therefore, we made a diagnosis of rupture of the left common iliac artery. During the angiography proce-

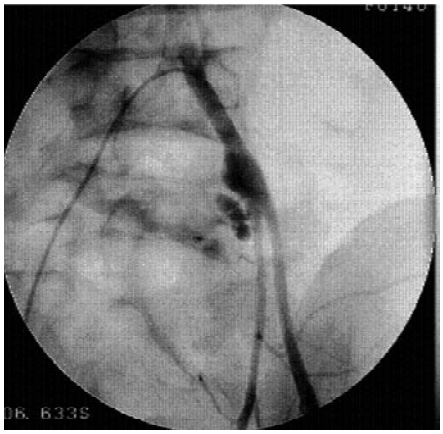

Fig. 2. Angiography image revealing extravasation of the left common iliac artery (B).

dure, the patient fell into shock, and was maintained by infusion and transfusion. He was quickly transferred to the department of vascular surgery, and emergency operation was performed.

The wall of the left common iliac artery and lower end of the aorta were both very weak, and the aorta could not be cramped and cut off at the lower end. On the right side, the common iliac artery was cut, while external and internal iliac arteries on the left were cut. To maintain blood flow to lower limb, we performed an axillo-femoral bypass (Fig. 3). Unfortunately, the patient died 3 days after the operation due to multiple organ failure caused by insufficient blood flow to the lower limb. A Pathological examination of the left common iliac artery revealed an arterial disorder caused by FMD (Fig. 3).

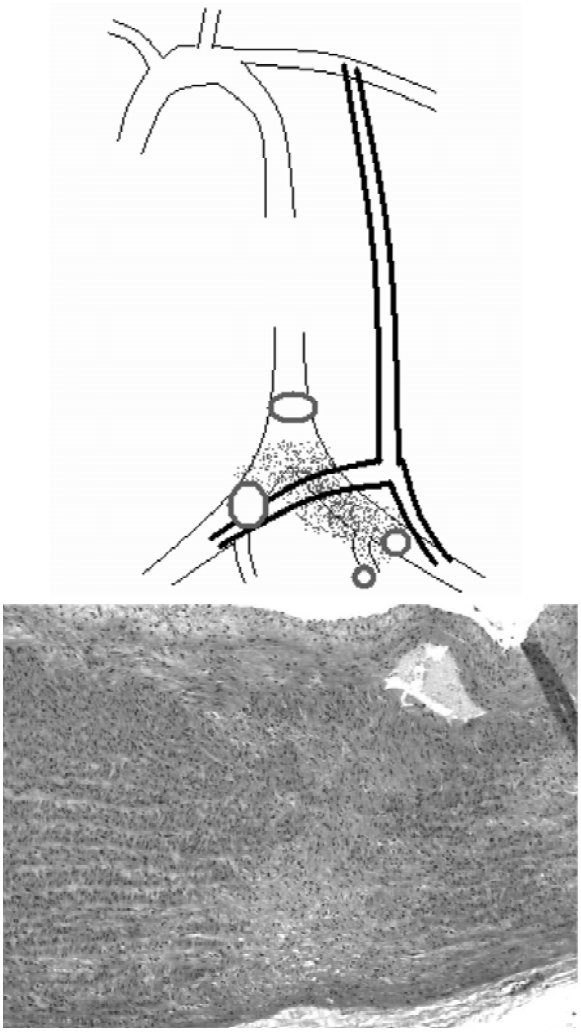

Fig. 3. A sketch of the present operation and pathological finding. Cracks in the smooth muscle of the artery are characteristic pathological findings in patients with FMD.

DISCUSSION

FMD is a non-atherosclerotic, noninflammatory vascular disease and, though a variety of genetic, mechanical, and hormonal factors have been proposed, the etiology of FMD remains uncertain. Although FMD most commonly affects the renal artery and internal carotid arteries, any artery throughout the body can be affected[1,5]. Most FMD cases are considered to be asymptomatic. However, an ischemic condition can arise in a distal area of an arterial disorder, depending on the region. In contrast, the cause of renovascular hypertension is well known, and is demonstrated by angiography a string-of-beads which is a result of stenosis in the artery[6]. Furthermore, secondary aneurysms, dissection and rupture can develop.

The lower extremity is a less common site for FMD to occur, as it is most often seen in the iliac arteries. Patients with lower extremity FMD may present with intermittent claudication, critical limb ischemia, or microembolism manifested as painful, cyanotic toes[7]. In this present case, no ischemic condition was present in the lower limbs and dissection and rupture occurred suddenly, before the patient had an opportunity to receive medical treatment. We were forced to treat the arterial rupture on an emergent basis without knowing the exact disease condition. Furthermore, we did not expect the arterial wall to be weak over a large portion prior to the operation.

Nine cases of iliac arterial dissections with FMD have been reported (Table 1). Most of those cases were presented as ischemia in the lower limbs without dissection or rupture, and lifesaving procedures such as iliofemoral bypass, interposition of the vein and conservative therapy, were performed for the patients. Honjo et al, reported a case of spontaneous rupture of the common iliac artery that developed in a 30-year-old previously healthy male, which is similar to the present case[7]. In that patient, emergency angiography revealed extravasation of the common iliac artery. Although an ileofemoral bypass was performed in an emergency operation, the patient died of circulation insufficiency. Generally, the operation mortality rate for patients with a rupture of an abdominal aneurysm exceeds 48%, which is nearly the same as in cases with a common iliac artery rupture[10]. When a rupture of the common iliac artery occurs, surgery is the only available

Table 1. Previously reported cases

Author (year)	Age	Sex	Presentation	Site of dissection	Treatment
Patel, et al. (1990)[3]	56	F	Hip and thigh claudication	Right common iliac artery	Conservative therapy
Thevenet, et al. (1992)[8]	45	F	Lower limb acute ischemia	Right external iliac artery	Aortabifemoral bypass
	51	M	Intermittent claudication	Both external iliac arteries	Bilateral iliofemoral bypass
	47	F	Abdominal pain	Local dissection in right external iliac artery	Bypass
	29	F	Iliac fossa pain	Left external iliac artery	Interposition with vein
	53	M	Intermittent claudication	Right external iliac artery	Iliofemoral bypass
Luck, et al. (2002)[9]	45	M	Intermittent claudication	Right external iliac artery	Thrombendarterectomy
Honjo, et al. (2004)[7]	30	M	Shock	Left common iliac artery	Iliofemoral bypass
Present case (2005)	21	M	Left abdominal pain	Left common iliac artery	Axillo-femoral bypass

treatment option. However, angiography is required for identification of the bleeding point and immediate reconstruction of blood flow is needed.

A number of patients with sudden lumbago caused by unbalance are seen at the general urology department. In such cases, it is important to bear in mind the possibility of retroperitoneal hemorrhage. If so, urgent treatment is needed, with angiography findings a key to successful therapy.

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和文抄録

尿管結石を疑う腰部痛にて発症した線維筋性異形成による総腸骨動脈破裂の1例

吉岡　　厳，有地　直子，徳川　茂樹
岸川　英史，西村　憲二，市川　靖二
兵庫県立西宮病院泌尿器科

線維筋性異形成による総腸骨動脈破裂の1例を経験したので報告する．症例は21歳，特に既往歴のない21歳の男性，突然の腰部激痛で発症した．当初，尿管結石による穿通発作が疑われた．腹部CTと血管造影にて総腸骨動脈の破裂による後腹膜血腫と診断された．緊急手術が行われたが，残念ながら患者は死亡した．病理学的検索により総腸骨動脈の線維筋性異形成が原因と診断された．非常に稀な症例であるが，突然の腰痛を主訴とする患者の中にこのような症例が含まれることを知っておくべきと考える．

（泌尿紀要 **53**：721-724，2007）